Botulinum Toxin for giant omphalocele abdominal wall reconstruction

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ABSTRACT

Introduction: The use of Botulinum Toxin A (BTA) has been shown to be safe and efficacious in neuromuscular blockade in both adult and pediatric patients. While BTA injections have been used safely in the pediatric population for a variety of medical conditions, its use in pediatric abdominal wall reconstruction has not been described. This report describes a unique surgical technique that will increase abdominal domain and allow for earlier closure of giant omphalocele defects.

Case report: A 33-week twin premie was born with a giant omphalocele. In an effort to achieve primary closure without the need for mesh, BTA injections were performed under ultrasound guidance two weeks prior to a planned closure. BTA injections included administration of 8 units at separate sites of the abdominal musculature bilaterally. After reduction, a component separation, and primary approximation of the fascial defect were achieved without signs of abdominal compartment syndrome.

Conclusion: BTA injection into the abdominal wall musculature provides a safe and effective mechanism to increase laxity of the abdominal wall musculature and decrease tension on the reconstruction for giant omphaloceles defects. The use of BTA may allow earlier repair in this subset of patients without the need for mesh.

The use of BTA injections as an adjunct in giant omphalocele repair appears novel. The following case reports describes a premature pediatric patient with giant omphalocele who underwent a joint operation with plastic and pediatric surgery in which BTA injections were used pre-operatively.

1. Introduction

The use of Botulinum Toxin A (BTA) has been shown to be safe and efficacious in neuromuscular blockade in both adult and pediatric patients for both aesthetic and reconstructive purposes. Of the reconstructive uses, BTA has been described as an adjunct in adult abdominal wall reconstruction. The theory behind its use is based on an increase in laxity of the abdominal wall musculature with a subsequent decrease in tension on facial defect repairs. While BTA injections have been used safely in the pediatric population for a variety of medical conditions including, but not limited to, hyperhidrosis, migraines, cleft lip, muscular hypertrophy, and torticollis [1], its use in pediatric abdominal wall reconstruction is not well described and is a novel technique. A *PubMed Medline* literature review failed to produce any reports using BTA in pediatric abdominal wall reconstruction.

The mechanism of action of BTA acts by inhibiting the release of acetylcholine, which prevents muscular contraction. The toxin takes effect within 10–14 days with a gradual decline in efficacy over 2–3 months. Furthermore, it has been shown to reduce inflammation and scarring due to an inhibition of glutamate and substance B release [1]. When administered into the abdominal wall musculature, the inhibitory effects of BTA work to increase the laxity of the abdominal wall, which leads to decreased tension on fascial repair sites [2]. In a single rat study, botulinum toxin use was shown to increase intraabdominal volume by 21%, which led to a subsequent decrease in pressure [3]. The effect of increased intraabdominal volume is useful in cases where post-operative intraabdominal pressures can be high, such as in patients undergoing omphalocele repair.

The approach to omphalocele repair is largely based on both size and associated comorbidities. Three different approaches have been described for the treatment of omphalocele: Immediate Repair, Staged Repair, and Delayed ("paint and wait") Repair. Immediate repair was largely favored in the 1800s, but gradually became less desirable with the use of staged repair in 1887 and the use of topical treatments to the

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omphalocele sac in 1899 [4]. Typically, a defect <5 cm without associated comorbidities are amenable to immediate repair. However, isolated omphaloceles are typically only found in 22% of cases [5], thus making a full systematic evaluation a necessity. Omphaloceles have been associated with pulmonary hypertension and hypoplasia, both disease processes that can make immediate primary repair less tolerable [5].

Staged repairs can be utilized with multiple staged operations, which can include silos and mesh that gradually reduce the extra-visceral contents [6]. Unfortunately, neurodevelopmental effects of subsequent exposure to multiple anesthetics, especially in children less than 3 years of age, have been hypothesized, however, these affect are currently unknown. The Delayed repair, or "paint and wait" approach, utilizes topical solutions (i.e. *Betadine*®, *Silvadene*®) to promote gradual coverage of the amnion in a process of escharification, followed by granulation and epithelialization. After sufficient tissue coverage of the amnion has been obtained, an interval repair is performed. The delayed approach has increased in popularity and is preferred in patients at high risk of abdominal compartment syndrome [4].

2. Case report

A 33-week male twin premie with intrauterine growth restriction was born with a giant omphalocele. After birth, he was followed by Pediatric Surgery for operative repair planning. Due to concerns for intraabdominal compression, poor weight gain, and the size of the defect, immediate surgical intervention was deferred and a delayed repair technique planned. During the neonatal period, the patient underwent dilute *Betadine*® paint to the omphalocele amnion membrane until gradual escharification and epithelialization occurred. This was followed by compression with an ACE wrap. Ultrasound at 17 months of age showed a 7.5 × 6 cm defect and 80% reducible giant omphalocele containing the liver. As the patient progressed with improvement in reducibility of omphalocele and weight gain and nutrition, plastic and reconstructive surgery was consulted for discussions regarding surgical planning of closure of the abdominal wall defect.

Secondary to concern for abdominal compartment syndrome (ACS) and the possible need for mesh, the reconstructive technique chosen included administration of BTA into the abdominal wall musculature to induce myofascial relaxation. "The toxin has a reversible paralytic effect that usually peaks about 1–2 weeks after injection, and neuronal activity begins to return about 3 months later." Our practice chose 14 days because of this theoretical peak in efficacy with exemplary mobility of the abdominal wall at 14 days post-injection [1]. Injections were performed under ultrasound guidance by Interventional Radiology two weeks prior to surgical intervention. Administration of eight units of BTA at three separate sites of each the superior, middle, and inferior aspects of the external, internal, and transversus abdominal musculature bilaterally was performed. A total of 150 units of BTA was provided. An interval examination two weeks after injections revealed a significant increase in the myofascial laxity of the abdominal wall, granting an easily reducible omphalocele (Fig. 1). Of note, the serious side effects of Botox which may include systemic affects or affecting areas away from the injection site including generalized muscle weakness, or problems swallowing, speaking, or breathing were not seen in our patient.

A combined surgical case was then performed by pediatric surgery and plastic and reconstructive surgery. Intraoperatively, the omphalocele was reduced and a longitudinal incision was made midline within the redundant tissue. The abdomen was entered and an extensive lysis of adhesions performed circumferentially from the pubic tubercle to the xiphoid. The liver and bowel were released from the omphalocele sac, as well as the anterior fascial sheath. The rectus abdominis fascia was entered bilaterally; the retro-rectus plane carefully opened; and the rectus muscles mobilized from the posterior sheath in a cephalad to caudad direction (Fig. 2).

Due to the myofascial laxity from the BTA injections, the anterior sheath fascia was able to be easily mobilized and reapproximated at the midline without the need for a bridging mesh. The anterior sheath was then closed with interrupted 2–0 Ethibond sutures in a figure-of-eight fashion. Peak airway pressures were monitored and noted to be within reference range with the abdominal wall completely closed. A complex closure of the skin was then performed utilizing 3-0 Vicryl sutures in a buried interrupted fashion in the Scarpa's fascia and deep dermis and 4-0 Monocryl suture in a running subcuticular fashion at the dermal-epidermal junction. Surgical glue was applied to protect the incision.

The patient was left intubated post-operatively and closely monitored in the pediatric intensive care unit to serially monitor peak airway pressures. On post-operative day number one, the patient was extubated

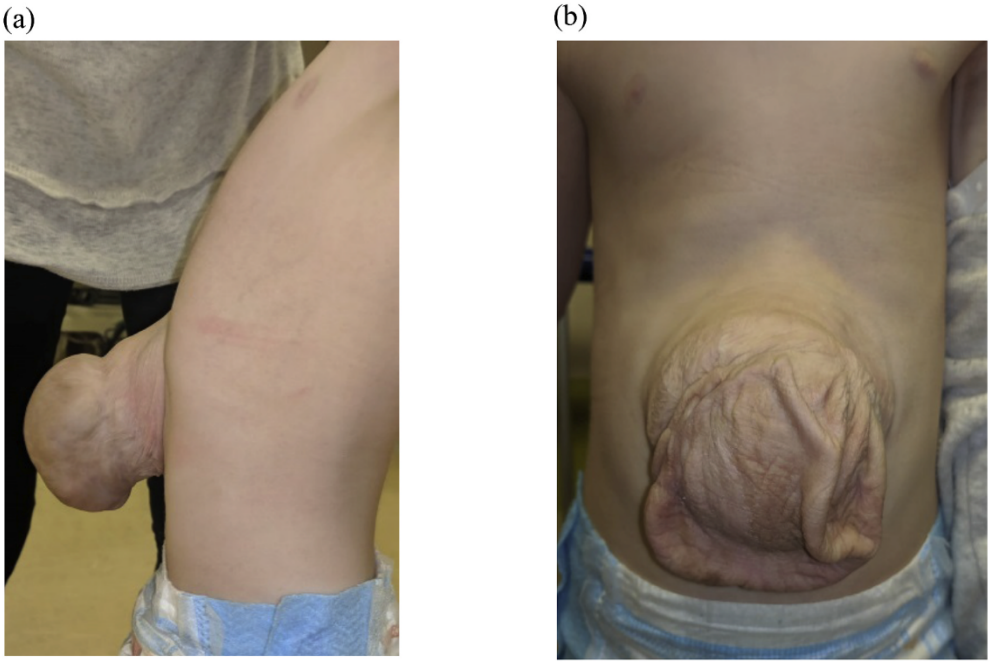

Fig. 1. (a–b): Omphalocele pre- and post-botox Injections(a) pre-botox injection (b) 2 Weeks post-botox injections.

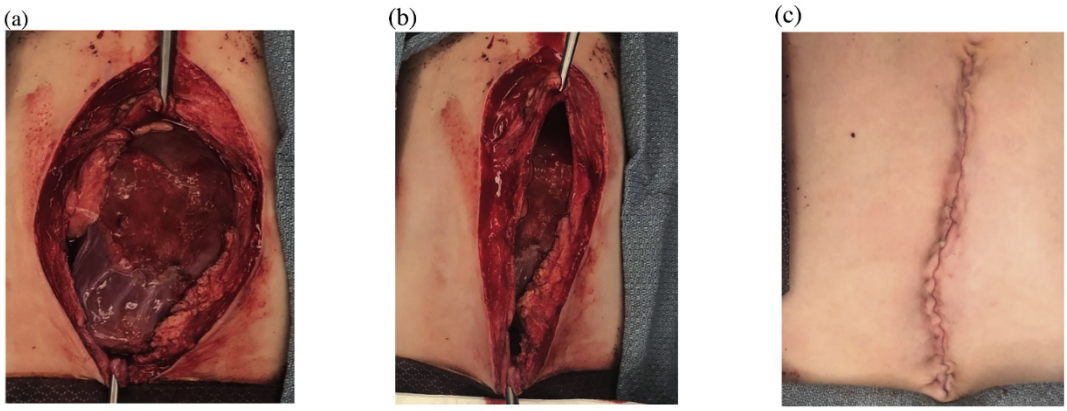

Fig. 2. (a–c): Intraoperative abdominal wall reconstruction & Closure(a) reduction of omphalocele contents (b) post-anterior component separation (c) post-closure.

and his diet slowly advanced. He was discharged home post-operative day number four. He was evaluated by both pediatric surgery and plastic and reconstructive surgery at 2 and 4 weeks post-operatively revealing a well-healed surgical scar, no evidence of recurrence, and a return to normal baseline activities (Fig. 3). Informal measurements of family satisfaction with repair and aesthetics were high.

3. Discussion

Omphaloceles have been categorized based on size and contents within the sac. Abdominal wall size defects of >5 cm and/or the presence of liver within the sac have been accepted as definition for a giant omphalocele. Various techniques have been described and performed, dating back to the 1800s. Even though techniques have evolved, primary reduction and closure remain technically challenging, particularly in patients born prematurely. Often these patients must undergo a delayed repair secondary to the need for medical optimization associated with prematurity and the concern for overall patient safety. This also provides adequate timing for thickening and epithelization of the omphalocele sac into a reducible ventral hernia.

When reduction and primary closure can be obtained, the concern remains high for the significant morbidity and mortality associated with the most feared complication of ACS. Strategies to monitor and prevent ACS after omphalocele hernia repair have been previously described [7], but the use of BTA to induce myofascial laxity to obtain primary closure has not been previously reported in the literature.

4. Conclusion

This case marks the first reported case of utilization of BTA as an adjunct in the pediatric population for obtaining primary closure of an omphalocele hernia defect without mesh. This technique is safe, simple, and efficacious. It is our hope that reporting our experience will add to the database of published literature, raise awareness of this technique, and allow of implementation for the treatment of giant omphaloceles.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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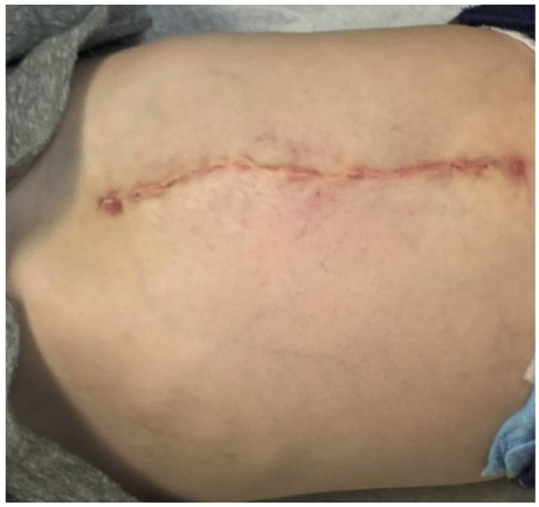

Fig. 3. Two Weeks Post-OperativePatient at two weeks after abdominal wall reconstruction without signs of recurrence.

Authorship

All authors attest that they meet current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

[1] Fu K, Teichgraeber J, Greives M. Botulinum toxin use in pediatric plastic surgery. Ann Plast Surg 2016;77:577–82.
[2] Motz B, Schlosser K, Heniford B. Chemical components separation. Plast Reconstr Surg 2018;142:58S–63S.
[3] Çakmak M, Caglayan F, Somuncu S, et al. Effect of paralysis of the abdominal wall muscles by botulinum A toxin to intraabdominal pressure: an experimental study. J Pediatr Surg 2006;41:821–5.
[4] Wagner J, Cusick R. Paint and wait management of giant omphaloceles. Semin Pediatr Surg 2019;28:95–100.
[5] Skarsgard E. Immediate versus staged repair of omphaloceles. Semin Pediatr Surg 2019;28:89–94.
[6] Bauman B, Stephens D, Gershone H, et al. Management of giant omphaloceles: a systematic review of methods of staged surgical vs. nonoperative delayed closure. J Pediatr Surg 2016;51:1725–30.
[7] Elsaied A, Medhat S, Sheir H, Aly K. The value of intra-abdominal pressure monitoring through transvesical route in the choice and outcome of management of congenital abdominal wall defects. Annals of Pediatric Surgery 2017;13:69–73.